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Case Report

Abdominopelvic inflammatory myofibroblastic tumor that metastasized to the vertebrae and liver: A case report and review of the literature

Sharon Kim [a], Jamie N. Bakkum-Gamez [b,*], Scott Okuno [c], Sarah Kerr [d], Sean C. Dowdy [b]

[a] Mayo Medical School, Rochester, MN 55905, USA
[b] Division of Gynecologic Surgery, Mayo Clinic, Rochester, MN 55905, USA
[c] Department of Oncology, Mayo Clinic, Rochester, MN 55905, USA
[d] Department of Laboratory Medicine and Pathology, Mayo Clinic, Rochester, MN 55905, USA

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Introduction

Inflammatory myofibroblastic tumor (IMT) is a rare mesenchymal tumor that is more common in children and young adults, with a median age of 9 years old and slight predominance in females (Unni and Hogendoom, 2002). Although IMT can involve any location in the body, the abdominopelvic region, retroperitoneum and lungs are most commonly affected (Coffin et al., 1995). Grossly, IMTs appear firm or fleshy with a white or tan surface and can resemble other benign or malignant tumors in appearance such as leiomyoma, leiomyosarcoma, and inflammatory liposarcoma (Rabban et al., 2005). Histologically, IMTs have spindle cell proliferation with inflammatory infiltrates of plasma cells, lymphocytes, and eosinophils (Gleason and Hornick, 2008). Immunohistochemical (IHC) staining will typically show reactivity for vimentin, smooth muscle actin and desmin (Unni and Hogendoom, 2002). Approximately 50% of IMTs will have IHC cytoplasmic positivity for anaplastic lymphoma kinase (ALK), which can be used to differentiate the tumor from the ALK-negative leiomyoma and leiomyosarcoma (Unni and Hogendoom, 2002; Rabban et al., 2005).

IMT is currently regarded as a neoplasm of intermediate biologic potential because of its tendency to locally recur but rarely metastasize (Coffin et al., 2007). Management of these tumors is challenging because there are no established protocols for primary or recurrent disease. Herein, we present a case of a woman whose abdominopelvic IMT was surgically debulked and treated with various chemotherapeutic agents, yet metastasized to the bone and liver, with extensive peritoneal sarcomatosis, within 15 months after diagnosis.

Case presentation

A previously healthy nulliparous 28-year old Caucasian woman presented to her primary care physician with a two-week history of postprandial abdominal pain. She was on oral contraceptives and had no family history of cancer. Physical examination revealed a palpable abdominal mass. The patient's CA 125 was 71 U/mL with negative AFP. Ultrasound and computed tomography (CT) scan showed ascites and multiple peritoneal masses (Fig. 1A), with the largest mass in the right upper quadrant measuring 6.5 × 8 cm. She was diagnosed with abdominal carcinomatosis and referred to gynecologic oncology.

Due to the uniform appearance of the masses, the differential diagnoses of disseminated leiomyomatosis and sarcoma were considered. The patient underwent an exploratory laparotomy which revealed extensive solid masses involving multiple viscera with impending obstruction (Fig. 1B). Tumor cytoreduction required resection of the sigmoid colon, two segments of ileum, the omentum, and the appendix. The masses were discrete and well-circumscribed, but deeply infiltrative into the adjacent muscularis of the small bowel and colon, prohibiting local excision without resection of the enteric tract. Despite the previously described resections to prevent obstruction and removal of several 8–10 cm masses from the ileal and jejunal mesenteries, several dozen nodules measuring up to 2 cm in size remained on the mesenteric border throughout the ileum and jejunum, prohibiting further resection. The uterus, ovaries and fallopian tubes were normal in appearance and left in situ as the patient desired fertility preservation.

Final pathologic analysis revealed the sigmoid colon, ileum and omentum to contain numerous nodules of IMT ranging in size from 0.5 cm to 13.5 cm in greatest dimension involving the serosa and mesentery (Fig. 1C). Histologically, the tumor was composed of a cellular proliferation of long, tapered, spindled cells forming loose fascicles in an edematous stroma with prominent lymphoplasmacytic infiltrate. There were occasional mitotic figures, including atypical forms. Some

* Corresponding author at: 200 First Street SW, Rochester, MN 55905, USA. Fax: +1 507 266 9300.
E-mail address: Bakkum.jamie@mayo.edu (J.N. Bakkum-Gamez).

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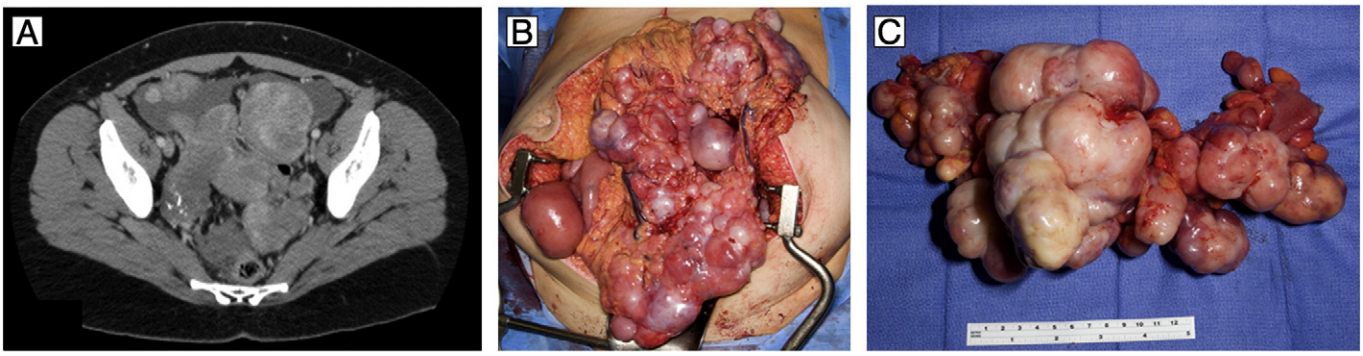

Fig. 1. A) Preoperative CT image demonstrating multiple solid peritoneal masses. B) Intraoperative findings of multiple solid masses involving pelvic and abdominal viscera. C) Resected sigmoid colon with numerous nodules of inflammatory myofibroblastic tumor ranging from 1.0 cm to 13.5 cm in greatest dimension involving the serosa and mesentery.

areas of tumor demonstrated ganglion-like round cells. By immunohistochemistry, the neoplastic cells were weakly reactive for smooth muscle actin (SMA) but negative for S100, desmin, KIT, anaplastic lymphoma kinase (ALK-1), CD34 and OSCAR-keratin, consistent with IMT (Fig. 2). The ascitic fluid and multiple lymph nodes from the omentum were negative for malignancy.

Postoperatively, the patient was started on prednisone for three months and experienced tumor progression. She was switched to imatinib for a month. During this time, the patient was noted to have significant pelvic pain and ureteral compression; she had bilateral ureteral stents placed with relief of symptoms. She was then started on liposomal doxorubicin; however, she had continued progression. The patient

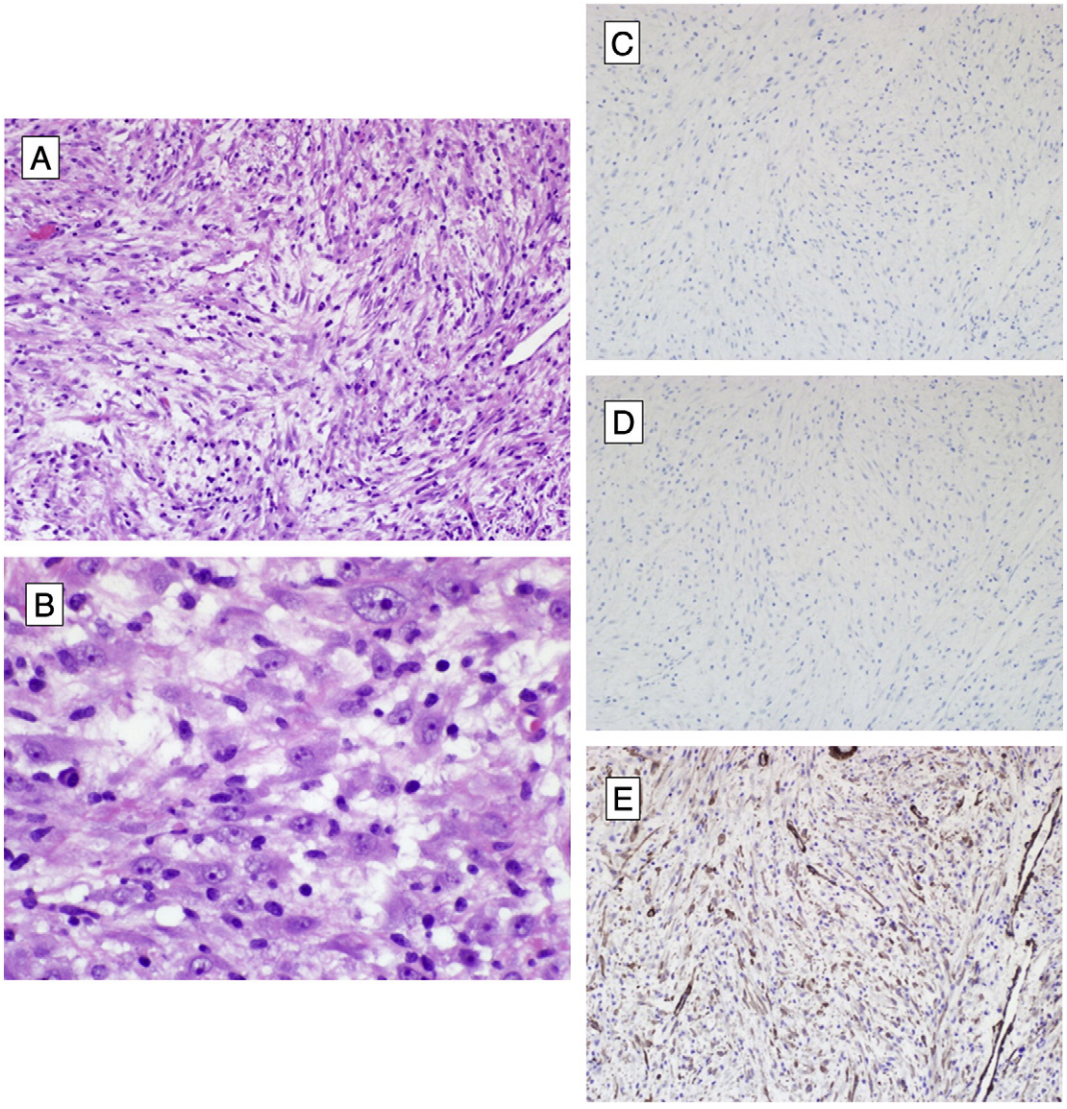

Fig. 2. Inflammatory myofibroblastic tumor. A) Hematoxylin and eosin stained preparation, 200× magnification, showing IMT with long, tapered, spindled cells arranged in loose fascicles in an edematous stroma with prominent lymphoplasmacytic infiltrate and occasional atypical mitotic figure. B) Hematoxylin and eosin stain, 600× magnification, demonstrating ganglion-like tumor cell atypia. C), D), E) Immunohistochemistry, 200× magnification, showing neoplastic cells to be negative for ALK-1 and desmin, but weakly positive for SMA, respectively.